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Giant sigmoid colon diverticulum

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ABSTRACT

Giant colonic diverticula are uncommon in all age groups and are especially rare within the pediatric population, with only one known previously reported case in a child. In this case presentation, we report a rare case of a pediatric GCD, review relevant epidemiology and histology, and discuss diagnostic and treatment options. The patient was an 11-year old female with worsening chronic constipation, abdominal pain, and vomiting. CT found a large pelvic mass that was removed via sigmoidectomy and found to be a Type 3 (true diverticula) GCD, which make up only 12% of all GCD's. Pediatric GCD's can be mistaken for the much more common enteric duplication cyst, but both pathologies require surgery. Because of the uncommon nature of GCD, the authors recommend CT as the diagnostic modality of choice because of its high sensitivity and ability to aid in preoperative planning. The authors also recommend treating pediatric GCD with segmental colectomy to avoid recurrence and even a remote risk of colonic adenocarcinoma.

1. Case report

An 11-year-old female with history of chronic constipation presented to the Riley Hospital for Children with intermittent abdominal and back pain and obstipation of approximately 2 weeks duration. The patient's past medical history was significant for multiple urinary tract infections and a current work-up for Osteogenesis Imperfecta given multiple fractures and lifelong constipation. The constipation had worsened over the previous 2–3 years resulting in 1–2 bowel movements a week. She took polyethylene glycol 2230 (Miralax®) daily. Several months prior to presentation, she had intermittent right lower quadrant abdominal and flank pain, dyschezia, and intermittent hematochezia. She presented to an outside hospital emergency department with dry heaving, bilious vomiting, right lower abdominal pain, and 1 week of constipation. Her physical examination was consistent with right lower quadrant abdominal tenderness without peritonitis. She had a leukocytosis and therefore a CT abdomen/pelvis was done to rule out appendicitis. The CT showed a significant stool burden and a large pelvic mass measuring 9 cm with mass effect on the bladder concerning for a colonic duplication or diverticulum [Fig. 1]. A few days of docusate, more laxatives, and magnesium citrate did not relieve the constipation and only after multiple enemas did she finally pass a small amount of stool. The following day she was transferred to Riley Hospital for Children at IU Health emergency department with right lower and now right upper quadrant abdominal pain. She had no fevers, no further vomiting, and other review of systems was negative. Laboratory workup in the Riley emergency department, including complete blood count, basic metabolic panel, and urinary analysis, was unremarkable.

The patient was admitted to the Pediatric General Surgery service and was taken to the operating room the following day given the imaging findings and ongoing abdominal pain. Proctosigmoidoscopy was normal to 15 cm and laparoscopy revealed a large, cystic, and mobile pelvic mass that was continuous with the sigmoid colon and was thought to be a duplication cyst or a large diverticulum [Fig. 2]. Given its size, the mass was surgically removed via partial sigmoid colon resection with primary anastomosis. Back table evaluation demonstrated a large sigmoid diverticulum full of inspissated stool with no surrounding inflammation. Pathology evaluation revealed a 7.2 cm long portion of colon with a large central diverticulum with dimensions of 5.8 × 5.4 × 4.5 cm. The diverticulum was noted to have a full wall thickness component and the diverticular mucosa was remarkable for some focal erythema and vague nodularity. The patient's hospital course was uneventful and she was discharged from the hospital 6 days later. Given unrelenting constipation, she later underwent colonic motility testing which demonstrated high amplitude propagated contractions with the administration of bisacodyl. For this reason, she underwent a Malone Antegrade Continence Enema procedure.

2. Discussion

Giant colonic diverticula (GCD), first described in 1946, are an

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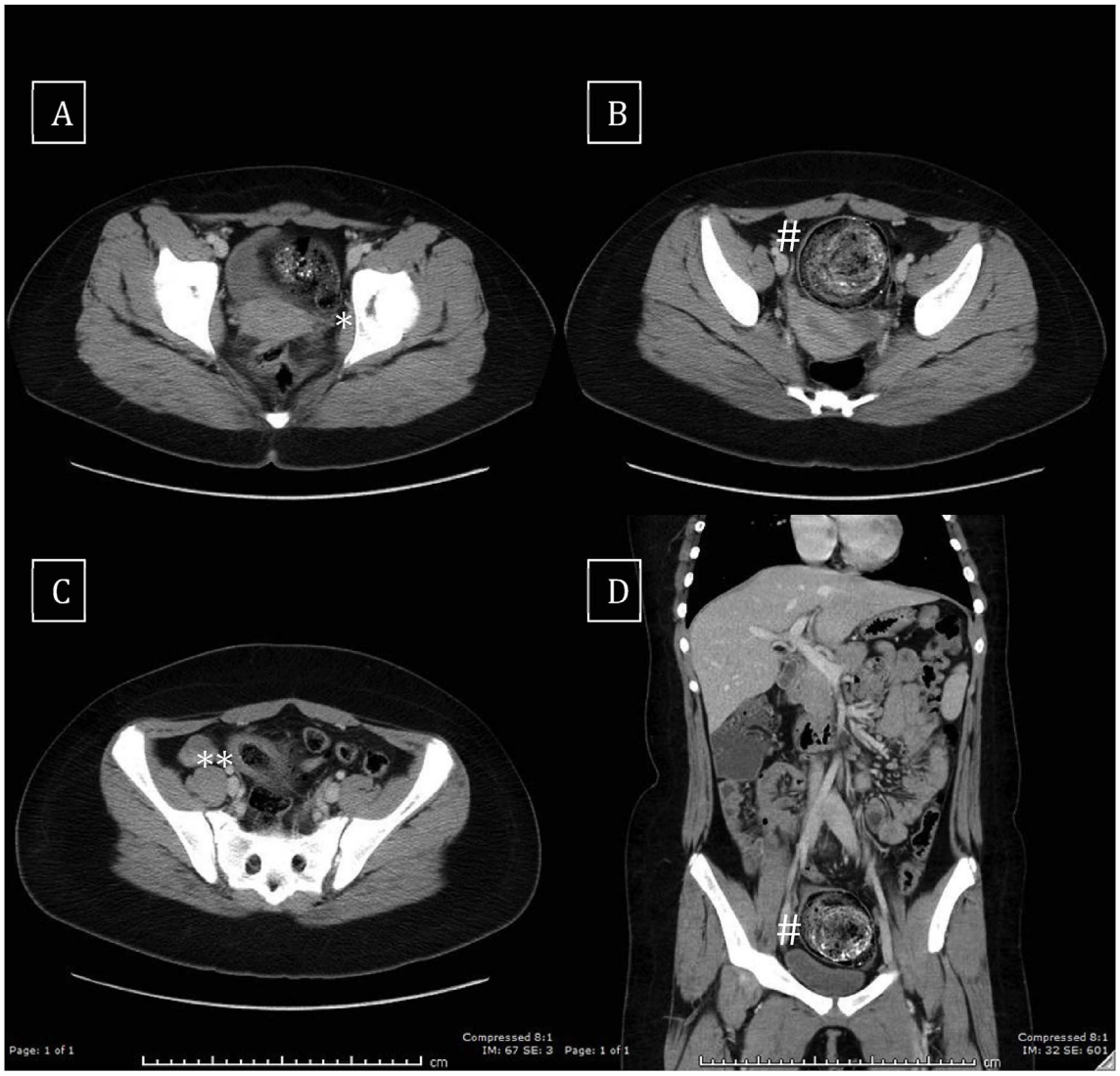

Fig. 1. CT images demonstrating the colonic diverticulum. Images A-C are axial CT images demonstrating the afferent sigmoid colon (*) and the efferent sigmoid colon (**) above and below the large sigmoid colon diverticulum (#). Image 4 demonstrates the diverticulum in the coronal plane.

Fig. 2. Laparoscopic image of the large sigmoid colon diverticulum in the pelvis. The normal caliber efferent sigmoid colon is at the bottom right of the picture.

uncommon pathology with diverse clinical presentations that have taken on many names such as “giant gas cyst”, “giant air cyst”, and “giant diverticulum”, but have now been given a standardized definition by Choong et al. as being a diverticulum in the colon that is 4 cm or larger [1,5]. GCD are quite rare in all age groups, with only 135 reported patients with GCD in the literature as of 2004, with a mean age between 60 and 79 [2]. Among this group of 135 patients, only 1 patient, an 8-year-old boy, was reported to be under 30 years old [3]. We were able to identify only one other possible case of a pediatric GCD found in a 12-year-old girl, although the authors of this case report didn't use the term “giant colonic diverticulum” and they also did not give an exact measurement of the diverticulum size so it is unclear if it would fit the current definition of a GCD [4]. We have presented a case of a GCD in an 11-year-old female.

This case is also notable because it represents a true diverticulum, an outpouching of the entire bowel wall thickness, and not the much more common false diverticulum, or pseudodiverticulum, which is a protrusion of the inner layers of the wall through a defect in the outer layers of the bowel. In 1988, McNutt et al. created a GCD classification system based on gross and histologic findings [6]. Type 1 GCDs (22% of Steenvoorde et al. patients) are pseudodiverticula in which the diverticular wall includes the muscular mucosa and the muscular propria, with an intact mucosa being present in the bowel [2,6]. Type 2 GCDs (66%) are inflammatory in nature and result from perforation of the mucosa resulting in an abscess cavity that communicates with the colonic lumen, with the diverticular wall including only reactive scar tissue and no portions of the bowel wall [2,6]. Finally, Type 3 GCDs (12%) are true diverticula that contains all the layers of the bowel in the diverticular wall [2,6]. While the male: female ratio was 1:1 in Type 1 and Type 2 GCDs, Steenvoorde et al. found that there was a 6:1 male:female preponderance for their Type 3 GCD patients [2]. Interestingly, our patient, which was found to have a Type 3 true diverticula, was a female.

In the Choong et al. classification system for GCD's of all ages, Type I GCD's are pseudodiverticula (87% of GCD's in their study) that are likely a rare complication of conventional diverticular disease, while Type II GCD's are true diverticula (13%) possibly related to a congenital communicating colonic duplication cyst [1]. During the workup of our patient, the primary two differential diagnoses were GCD and duplication cyst. Enteric duplication cysts are developmental anomalies with an estimated incidence of 1:4500 births, occurring anywhere in the gastrointestinal tract and are typically diagnosed within the first 2 years of life [7–9]. Only 13% of duplication cysts occur in the colon and they are often indistinguishable on CT from GCD, as was the case in our patient [10]. For surgeons in such a diagnostic dilemma, the authors of this paper advocate for surgery, as the management of both a GCD and a symptomatic duplication cyst is operative removal.

By far, the most common presenting symptom of GCD's is abdominal pain (68% of cases) [2]. Our patient had abdominal pain but also had constipation, emesis, and hematochezia (present in 18%, 12%, and 8% respectively) [2]. Interestingly, Choong et al. found that in their patient population, only 9% of the Type I (pseudodiverticula) patients experienced constipation, while 25% of the Type II (true diverticula) had constipation [1]. Constipation is a common symptom in the pediatric population in general and the authors believe that in this patient, constipation was likely a predisposing factor in the formation of her true diverticulum. Regardless, we can confidently assume that the GCD was not the sole causative factor in this patient's constipation, as five months post segmental colectomy, our patient was still struggling with constipation. Of note, this does differ from the experience of the 8-year-old boy in the Barlow et al. paper, whose chronic life-long constipation was apparently cured after the removal of the GCD [3].

Because of the varying clinical presentations and the rare nature of GCD, the diagnosis is primarily radiologic. Ultrasound and colonoscopy have historically not been considered to be very helpful in diagnosing GCD [2,11], and while barium enema's have frequently been used and can show the presence of communication, they have been associated with the risk of perforation of the GCD [12–14]. Steenvoorde et al. found that the diagnosis of GCD can accurately be made with plain film X-rays, as 103/104 GCD X-rays showed the classic gas filled structure [2]. They did however, mention that 17 out of 17 GCD's were successfully diagnosed with the CT scanner and that this imaging modality seemed to be more informative in providing the number of GCD's [19], the presence of a communicating GCD even when not picked up on barium enema [15], and the presence of any complications not otherwise noted on X-ray [16]. There are currently no known recommendations on the workup for GCD's in the pediatric population. In 1975, Barlow et al. successfully performed a barium enema and proctoscopy with biopsy on their 8-year-old patient. In the present case, CT was successfully used. The authors believe that in the modern era, the CT scan is the imaging modality of choice for its high sensitivity, ability to provide the most information for preoperative planning, and lack of complication risk found with other imaging modalities.

Complication rates of GCD range from 28 to 35% and have been reported to be as high as 58% in patients with true diverticula, and thus surgical removal is pertinent [1,2]. Choong et al. recommends a treatment algorithm based on the condition of the colon near the GCD, in which diverticulectomy is performed for healthy colon and segmental colectomy is performed for colon that is inflamed, necrotic, or has significant diverticulosis in order to both remove the GCD and to also prevent future diverticular disease [1]. Historically, approximately 15% of patients have received diverticulectomy and 72% have received colonic resection [2]. Steenvoorde et al. support performing a segmental colectomy with possible protecting colostomy for all GCD patients, citing cases of recurring GCD after diverticulectomy alone [17] and cases of GCD complications of acute abdomen and adenocarcinoma [18]. The fact that most GCD's have a large neck makes them unlikely to result in diverticulitis, which requires the enclosure and inflammation a fecolith. However, because of the significant risk of other complications from GCD noted above, the authors' do agree that surgical removal of GCD is important. In this case, a segmental colectomy with primary anastomosis was performed successfully and the authors believe that colectomy is the best surgical option.

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Conflicts of interest

None.

Clinical trial registration

Not applicable.

Contributors' statements

Christian Corbin Frye: Dr. Frye completed the relevant literature review and helped to author and review the initial and final manuscripts.

Matthew Paul Landman: Dr. Landman saw and operated on the patient in this case report. He conceptualized the manuscript and was instrumental in the writing and editing of the initial and final manuscripts.

All authors approved the final manuscript and agree to be accountable for all aspects of the work.

Table of contents summary

We present a pediatric patient with a giant colonic diverticulum and discuss relevant epidemiology, symptomatology, and diagnostic and treatment modalities.

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